GIANT SOLITARY OSTEOCHONDROMA OF THE PROXIMAL HUMERUS TREATED BY RESECTION AND FIBULAR AUTOGRAFT RECONSTRUCTION

by M. GEBHART, J. VADOUD SEYEDI and F. LEJEUNE

A giant solitary osteochondroma of the proximal humerus in a 9-year-old boy was widely resected. Reconstruction of the bone defect was achieved with two nonvascularized fibula segments which gave sufficient stability to the proximal humerus and yielded rapid bone remodeling into an almost normal cortical bone after six months.

Keywords : osteochondroma ; bone tumor ; fibula graft.
Mots-clés : ostéochondrome ; tumeur osseuse ; greffe péronière.

RÉSUMÉ

M. GEBHART, J. VADOUD SEYEDI et F. LEJEUNE. Ostéochondrome géant solitaire de l'extrémité supérieure de l'humérus, traité par résection et reconstruction à l'aide d'un greffon péronier autologue.

Un ostéochondrome géant solitaire de l'extrémité supérieure de l'humérus fut réséqué largement chez un enfant de neuf ans. La reconstruction de la perte de substance osseuse fut réalisée par deux segments non vascularisées de péroné. Le montage assura une stabilité osseuse primaire satisfaisante et l'intégration de la greffe osseuse s'effectua durant les six premiers mois postopératoires. Le patient a récupéré une fonction normale de l'épaule opérée.

SAMENVATTING

M. GEBHART, J. VADOUD SEYEDI en F. LEJEUNE. Gigant solitair osteochondroma van het proximale uiteinde van de humerus, behandeld met resectie en reconstructie d.m.v. een fibulaire ent.

Een gigantisch osteochondroma van het proximale uiteinde van de humerus werd volledig verwijderd bij een 9-jarig kind. De reconstructie gebeurde d.m.v. 2 niet gevasculariseerde fibulaspanen. Er werd primair een bevredigende stabiliteit bekomen. De assimilatie van de botenten gebeurde in de loop van de eerste zes postoperatieve maanden. De funktionele recuperatie van het schoudergewricht was volledig.

INTRODUCTION

Solitary osteochondroma (SO) is the most common benign tumor involving the skeleton. It appears as a painless enlargement commonly found within the metaphysis of a long bone. Most are discovered during the three first decades of life (8). The majority of the SO are asymptomatic and are discovered only when symptoms occur. Symptoms consist of pain, limitation of joint motion, edema, paresia or paresthesia due to compression of neurovascular structures. Local pain may be explained also by the presence of bursitis, which covers the cup of the osteochondroma. Involvement of the proximal humerus by an SO is observed in about 25% of the patients (8). We report on a case of a giant SO which appeared with major impairment of adduction and internal rotational motion of the arm. Extensive resection of the tumor was followed by reconstruc-

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tion of the bone defect using two nonvascularized fibula segments.

CASE REPORT

The patient was a 9-year-old male who reported over a 3-year period the history of an increasing swollen mass in the proximal metaphyseal region of his left humerus, which had been growing more rapidly 5 months prior to consultation. At this time the severe limitation of adduction of the left arm prevented normal childhood activities (spontaneous abduction of 70°).

On physical examination, we observed a hard, lobulated mass in the posteromedial area of the proximal humerus. The tumor was clearly visible on external rotation. The functional impairment consisted of spontaneous adduction of 70° and totally limited internal rotation (0°) of the shoulder joint. There was no neurological deficit. The humeral vessels were palpable and displaced anteriorly within a groove of the tumor. No sign of veinous stasis was observed.

The radiographs showed a giant exophytic tumor arising from the proximal metaphyseal region of the left humerus (fig. 1 a, b). CT scan confirmed the presence of a large bone tumor without any soft tissue mass. Moderate pathological uptake in the tumoral area was seen on the technetium bone scan.

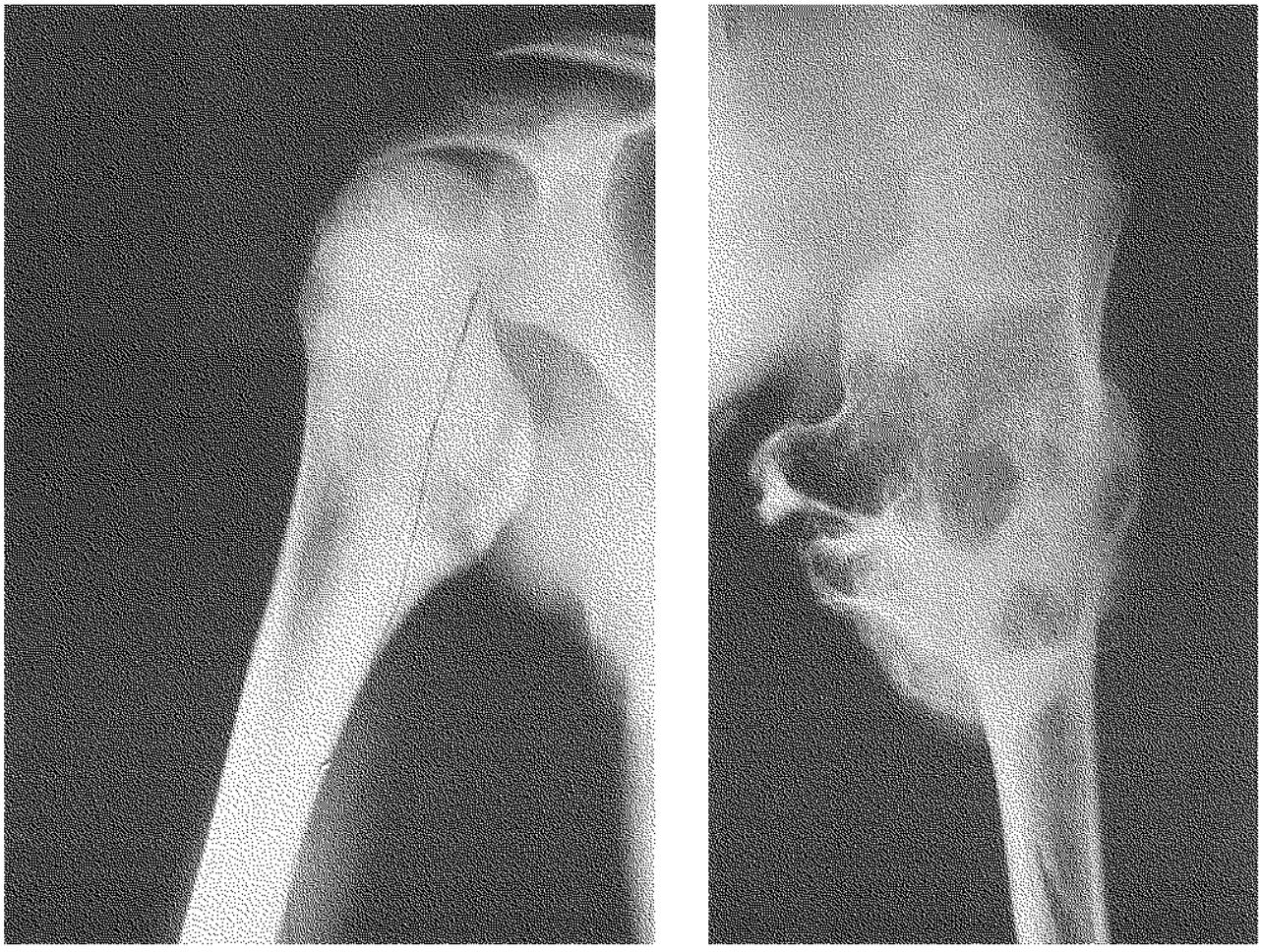

Fig. 1a *Fig. 1b*

Fig. 1. — a) Osteochondroma of the proximal humerus 3 years prior to treatment. — b) Three years later.

The preoperative diagnosis suggested by the xrays and clinical examination was either a benign giant osteochondroma or a chondrosarcoma arising from an osteochondroma. An open biopsy was performed, and histology revealed a benign osteochondroma. One month later, the definitive surgical procedure was done.

The tumor was exposed by a delto-pectoral approach which was prolonged distally in the medial part of the arm. First the neuronal and the vascular structures were separated from the tumor and the tumoral borders delimited. Then the deltoid and brachial muscles were transsected in part. This allowed clear exposure to the base of the osteochondroma. Multiple drill holes were placed around the base of the tumor. These drill holes were connected to each other using an osteotome. This technique avoided a possible fracture through the proximal humerus. With mild traction, the tumoral mass was freed from the surrounding tissues with preservation of the proximal metaphysis. After removal of the tumor, the humeral head was connected to the humeral diaphysis by a very small bone bridge. A non-vascularized fibular bone graft was taken from the left leg. The graft was divided into 2 pieces and placed in the medullary cavity of the humerus and maintained by 3 cerclages (fig. 2). The surgical wounds were closed with an aspiration drain left in place, and the arm was protected with an over-the-shoulder arm brace. No surgical complications occurred, and passive motion was started on the third postoperative day. Active shoulder exercises were allowed after a 6-week period.

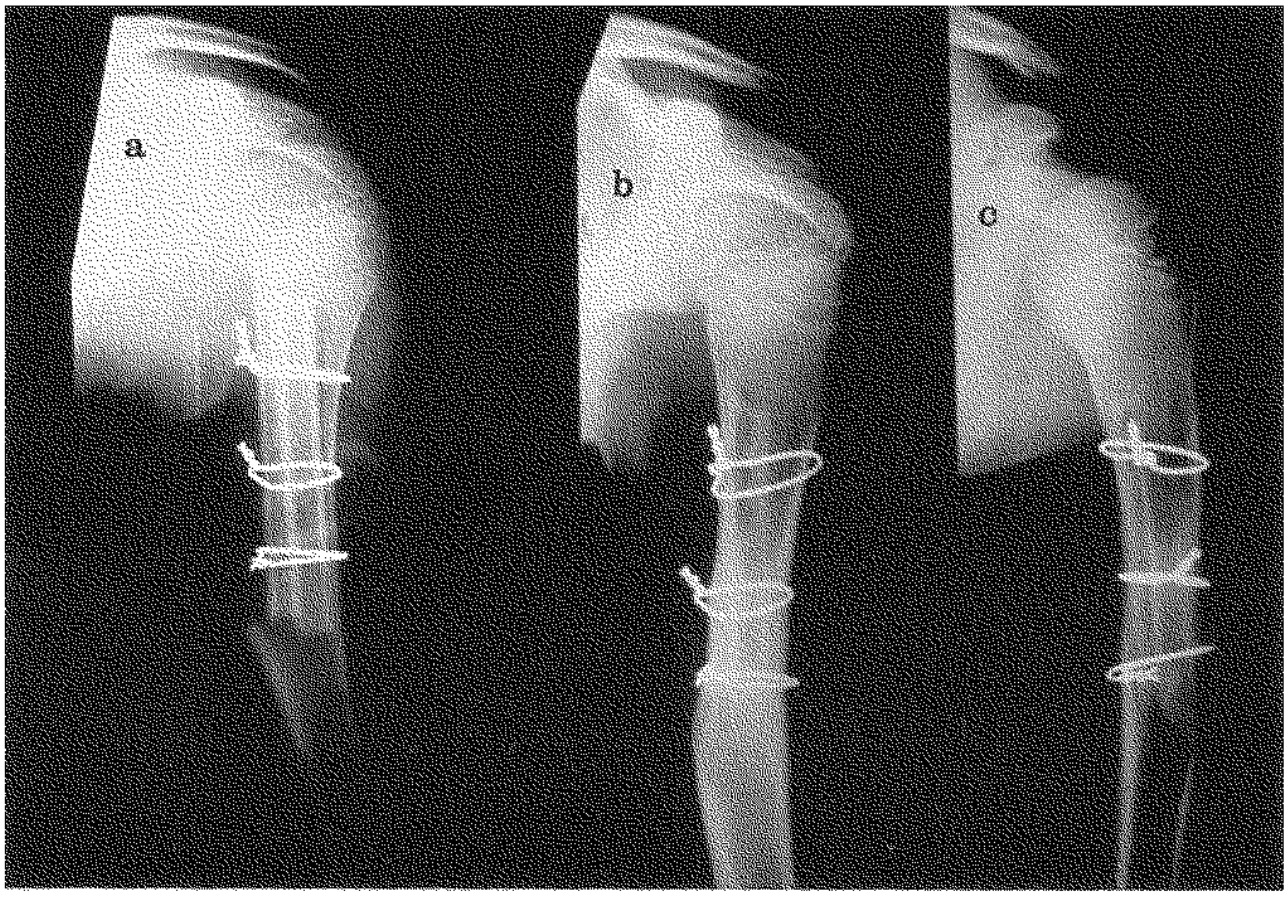

Fig. 2. — Proximal humerus :
a) After resection. Reconstruction by two fibula segments. — b) c) Six months later.

Radiographs taken one year postoperatively showed almost complete resorption of the two fibula segments with bone remodeling of the proximal humerus. Cortical bone reformed around a medullary cavity. No local recurrence of the tumor has been seen so far. The patient has normal use of his upper extremity without restricted range of motion as compared to the healthy opposite upper extremity.

DISCUSSION

The solitary osteochondroma is a cartilaginous benign tumor. It represents 50% of all the benign bone neoplasms and 10 to 15% of all the primary tumors (8).

The lesion is usually discovered during the second decade of life with a male-to-female ratio of 2 : 1 (12). SO appears as a lobulated, firm mass with a long pedicle. A large base of implantation of the SO as in this case should be biopsied before resection because a chondrosarcoma can give a similar appearance. The tumor is composed of histologically normal bone, containing bone marrow, and coated partially by benign cartilage. The two most common sites are the distal metaphysis of the femur and the proximal metaphysis of the tibia which together represent 50% of the cases (8). Other sites of predilection are the proximal metaphysis of the humerus, the distal end of the radius and the distal metaphysis of the tibia. Localizations such as foot (3, 7), scapula (2), spine (14) and even mandible (1) have been reported. The proximal metaphysis of humerus is usually affected by SO in the posterointernal part.

The usual clinical sign is a painless mass situated near a joint (4). When compression or irritation of an adjacent structure arises, the tumor becomes painful because of either tendinitis or bursitis (13). With the development of the tumor, limitation of joint motion is generally observed and in the case of SO of the proximal part of the humerus, a reduction in internal rotation and adduction frequently occurs. In cases of an SO within the popliteal space, swelling of the lower extremity by popliteal vein compression or by pseudoaneurysm formation of the popliteal artery have been described in the literature (11).

The tumor occurs only in bones developed from cartilage (enchondral ossification), mostly in the long tubular bones. Some authors (5, 10) consider this tumor as an abnormal development of a cartilage island displaced from an altered periosteum which gives rise to osteocartilaginous exostoses. But according to others (9), SO is caused by a hyperplastic process, the consequence of a congenital abnormality of blood vessels. The pathogenesis of the tumor has been considered as a defect of the "ring of Rosier" (fibrous tissue surrounding the epiphyseal plate). In this hypothesis, cartilage protrudes through the ring of Rosier. This cartilage island migrates with the newly formed bone and gives rise to bone formation by enchondral ossification. If there is a major defect of the ring of Rosier, mostly lateral bone growth will take place. The microscopic appearance consists of a cartilage cap covering trabecular bone. This hyaline cartilage is cellular with the chondrocytes arranged in vertical rows. Binucleate chondrocytes are frequently observed (8). During the normal growing process, the cartilage regresses progressively and is replaced by a layer of bone, but residual microscopic foci of cartilage may be identified in adult bone (8).

Accelerated growth of the SO occurs during puberty, and the growth of the tumor usually stops at the end of skeletal maturation. Subsequent enlargement during adulthood should give rise to a high suspicion of sarcomatous degeneration (most frequently around the coxofemoral joint) but this event occurs only in 1% of all cases (6). However, pregnancy and lactation may stimulate new growth that mimics malignancy.

Operative treatment is indicated when the tumor becomes a source of inconvenience. The classic treatment is complete excision of the osteochondroma at its base. Sometimes, especially in the lower extremity, bone grafting as well as stabilization of bone is necessary to avoid postoperative fractures.

The most important principle in the treatment of patients with osseous neoplasia is accurate histological diagnosis. Indeed, biopsy is essential to avoid the undertreatment of a malignant neoplasm or to avoid the overtreatment of a benign lesion. In our case, after confirmation of a benign SO,

a surgical technique had been chosen : 1) to allow a wide en bloc resection ; 2) to maintain normal growth of the humerus by conservation of the epiphyseal plate ; and 3) to restore sufficient stability by reconstructing the metaphyseal bone defect by an autogenous bone graft in the form of two nonvascularized fibula grafts.

Regular clinical examination and serial radiographs have revealed no signs of recurrence, and normal shoulder motion has been restored.

A similar surgical technique had already been described. In a series of five patients (15), autogenous corticocancellous tibial bone graft was used and the lower extremity was immobilized for weeks. As removal of large amounts of tibial bone could lead to iatrogenic fractures, our preferred approach is to use fibular grafts in order to avoid this inconvenience. By fixing these bone grafts to the humeral head and the proximal humeral diaphysis, better primary stability at the site of implantation should be obtained. No cast immobilization of the lower extremity was necessary and function became normal after 2 months.

CONCLUSION

This paper reports on a giant osteochondroma of the proximal humerus which was widely resected. Stabilization of the bone defect was achieved by interposing two fibula segments. This yielded good stabilization of the proximal humerus. Six months after implantation the bone grafts have already been reintegrated into the proximal humerus with radiographic remodeling of the grafts into cortical bone containing a medullary cavity. Normal function and no sign of local recurrence have been observed after 30 months.

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